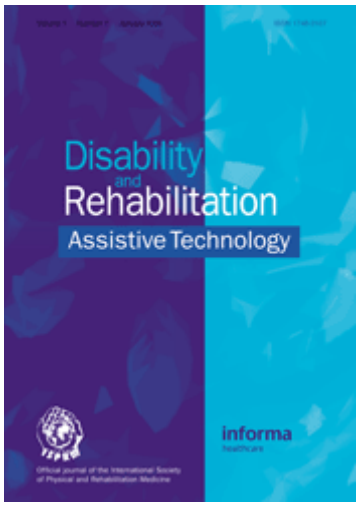

Experience of Telehealth in People with Motor Neurone Disease using Non-Invasive Ventilation

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Implications for Rehabilitation

- Care for people with MND demands a flexible approach to accommodate the diversity of clinical needs and relentless physical deterioration
- Telehealth allows clinicians to provide person-centred care for everyone with MND through frequent monitoring
- Holistic and rehabilitative service facilitated by telehealth is generally acceptable and preferred to routine appointments among MND NIV patients
- Telehealth promotes time efficient engagement with professionals that leads to symptom awareness and interpretation, while benefiting physical and psychological well-being of MND NIV patients

Experience of Telehealth in People with Motor Neurone Disease using Non-Invasive Ventilation

Abstract

Introduction: Evidence is emerging that telehealth provides timely and cost-effective support for individuals with motor neurone disease (MND). However, little is known about the subjective experience of using telehealth. This study was designed to examine the experiences of using telemonitoring in patients with MND on non-invasive ventilation (NIV).

Methods: Semi-structured interviews were conducted with seven patients (five males; mean age=63yrs; median illness duration=14m), who used a telemonitoring device for 24 weeks. Caregivers were present at five of the interviews; they supported communications and provided their feedback. Interviews were audio recorded and transcribed verbatim. Thematic analysis was conducted to find overarching themes.

Results: Five themes were identified: Benefits of Timely Intervention, Reducing the Unnecessary, Increased Self-Awareness, Taking Initiative, and Technical Challenges. Overall, timely interventions were observed as a result of regular monitoring, contributing to both physical and psychological well-being of the participants. The patient-caregiver dyads suggested that telemonitoring could reduce costs, save time and ameliorate hassles associated with attending hospital appointments. Participants articulated that telemonitoring enabled symptom awareness and interpretation; the device also enabled the participants to raise concerns and/or requests to the healthcare professionals via the messaging system. Participants confirmed that the telemonitoring device was easy to use, despite some technical issues.

Conclusions: Telemonitoring was positively experienced. The findings suggest this approach is empowering and effective in promoting patients' well-being, while potentially reducing unnecessary clinical contact.

Keywords: Telemonitoring; Telehealth; Qualitative study; Non-invasive ventilation; Motor neurone disease; Amyotrophic lateral sclerosis

Introduction

The United Kingdom National Institute for Health and Care Excellence (NICE) [1] recommends flexible approaches to the care of patients with motor neurone disease (MND) – also known as amyotrophic lateral sclerosis (ALS) – according to their needs. MND is a fatal neurodegenerative disease which primarily impacts on the motor systems. Appropriate symptom management, particularly of respiratory challenges, is reflected in longer survival rates [2]. To this end, and to improve quality of life for patients, the NICE guidelines prompt clinicians to consider: an individual's capability to attend the clinic, the appropriate frequency for their assessments and arrangements for prompt review when significant deterioration is declared. These recommendations reflect the diversity in symptomatology and prognosis [3], the relentless deterioration, and rapid progression of the condition towards the final stage [4]. The guideline also suggests the need for a new approach to meet the challenging task of accommodating the diversity of clinical needs.

In view of the NICE guidelines, and the use of telehealth emerging as a promising platform for clinicians to provide flexible approaches, we developed a telemonitoring system, using the Docobo^{Ltd} Careportal® device. The device allows clinicians to monitor their service users more frequently, outside of the clinic, regarding their symptom changes, non-invasive ventilation (NIV) related issues, nocturnal blood oxygen saturation levels, and patient-ventilator interaction data. Careportal® also incorporates a messaging system for patient-clinician communications. The details of system and intervention protocol are reported elsewhere [5].

There are a few studies that have examined telehealth systems in the care of MND. These include videoconferencing [6], nurse-led weekly telephone call follow-up [7,8], and weekly telemonitoring of NIV compliance [9,10]. Findings suggest that these telehealth systems are effective and provide an approach for meeting the NICE recommendations to tailor care for each patient. Nevertheless, the subjective experience of *using* telehealth must be explored to ensure that any changes to MND care procedures are not made at the expense of patients. Little is known of the advantages and disadvantages of telehealth for MND patients themselves. As far as we are aware, the only available report of subjective accounts of using a telehealth service amongst people with MND is based on a satisfaction questionnaire conducted via telephone [7]. There has been an attempt [11] to co-design a MND telehealth service with MND patients and carers; however, at the consultation meetings potential users of telehealth could only relate to aspirational benefits because at that time, none of the MND patients or their carers had used Telehealth.

Amongst other illness groups (e.g. diabetes and chronic heart failure), there are suggestions that telemonitoring is supportive [12-19]. Patients generally found few problems using telemonitoring, and whilst some difficulties were reported – with either the telemonitoring technology or internet connectivity – these were minor [13,15,17]. Reports referred to a sense of reassurance and peace of mind through using telemonitoring system, which were attributed to perceived continuous monitoring and support by professionals [12,13, 18]. Also, telemonitoring has been linked with increased knowledge of one's condition [12,18,19] and thereby facilitating patients to identify the 'norm' of their symptoms [12,19]. Although an increased awareness of symptoms may have a negative effect on the minority of patients [14,18], self-awareness of health status through telemonitoring was generally perceived to be positive and empowering to patients [12]. Hanley et al. further reported that telehealth promoted healthy behaviour, although they acknowledged that this may have been facilitated by participants' awareness of their data was being monitored [15]. Telemonitoring was seen to be a more convenient approach for patients, and the need for less appointments with healthcare professionals was appreciated [15,18].

Despite these positive findings of using a telehealth system, we cannot assuming subjective accounts of the benefits of telemonitoring in other chronic conditions will generalise to the MND population; there are clear differences in symptoms and prognoses of other conditions compared to MND. Thus, the current study was designed to explore subjective experiences of telehealth as facilitated by the Careportal® device as a regular monitoring service amongst people with MND who were using NIV.

Methods

Ethics

Ethical approval to conduct this study was obtained from United Kingdom National Health Service (NHS) Research Ethics Service (Ref no. anonymous) and the Research and Development department of the study site.

Potential participants were given an information sheet about the study and written consent was obtained after participants had a chance to ask questions about the study and their role in it.

Participants

Opportunity sampling was employed to recruit patients who had completed a 24-week trial of telemonitoring using Careportal® [5]. 13 patients with MND on NIV were recruited for the 24-week trial. Of the original 13, three patients died before the completion of the trial and an additional three before interviews were conducted. The remaining seven patients agreed to be interviewed about their experience of care associated with telemonitoring. Table 1 shows demographics and illness characteristics of the seven patients at the time of the trial. The median illness duration was 14 months and the median time on NIV was 12 months. The average time between the end of trial and interview was 8 months.

Five carers were also recruited to explore their experiences of using the Careportal®. An interview with the other two carers was perceived unnecessary by the patients because they had little

involvement during the trial. Joint interviews were preferred by patients and their carers thus an arrangement was made to interview them together.

[Table 1 about here]

Data Collection

Semi-structured interviews were conducted by a non-clinical researcher, who monitored the weekly data input during the 24-week trial. Interview questions (Appendix 1) were developed by the research team to explore subjective experiences of using Careportal® and to understand the impact, if any, of having a telemonitoring service in the management of their MND illness situation. All questions were addressed to both patients and their carers with one extra question only applicable to the carers.

All the interviews were conducted in participant's homes. During the interview, John and Margaret used a communication device (i.e. iPad and an eye control computer), and three patients (i.e. John, Margaret, Mary) required communication support from their carer. Carer's interpretations of patient's comments were always checked with the patient. The length of interviews ranged between 30 minutes and 50 minutes with an average duration of 35 minutes. Interviews were audio recorded and later transcribed verbatim.

Data analysis

A qualitative study is explorative and researchers aim to understand subjective experiences of a particular phenomenon by immersing themselves in the data and – through an iterative analysis process – make sense of the narratives. As argued by Chamberlain, qualitative studies are interested in what underlies the surface to answer 'how' and 'why' questions [22]. Thematic analysis [23] was employed to answer 'how' telemonitoring service was perceived by the participants and 'why' it was so. Codes were inductively generated from the transcripts and they were later examined to identify the overarching themes, which were subsequently assessed to ensure that they reflected the data

set. Initial coding of the transcripts was assisted by software for qualitative analysis (NVivo11). The analysis was conducted by the same non-clinical researcher who interviewed the participants. The credibility of the analysis was examined by the research team; the findings were presented at a team meeting, and a list of codes (Appendix 2) and all transcripts were shared with the team for a credibility examination.

Results

Thematic analysis identified five themes: benefits of timely intervention, reducing the unnecessary, increased self-awareness, taking initiative, and technical challenges. The following section expands each theme using supportive quotes from the narrative accounts listed in brief sequential tables 2-6. Each quote is cross referenced from the table into narrative of interpretation. Three full stops in brackets indicate an omission of words.

Benefits of timely intervention

Timely interventions were observed by six of the seven patients and all caregivers as a result of regular monitoring of clinical information via Careportal®, contributing to both physical and psychological well-being. Its benefits on physical well-being were observed when oximetry results prompted interventions (1)a. Whilst participants James and Charlie did not perceive any gain from Careportal® in terms of physical well-being at the time of trial, they still acknowledged potential benefits for the future (1)b.

In addition to clinical benefits from using telemonitoring, a positive impact on psychological well-being was reported. Participants appreciated their data being monitored by professionals who would make timely actions if they saw any irregular signs: (1)c. The significance of these positive impacts of Careportal® on psychological well-being was further highlighted by two participants who had identified psychological well-being to be the key to coping with the condition (1)d.

Whilst the benefits of timely intervention were expressed as above, John and his professional home carer did not perceive any benefits because of the good home care support he was receiving, as argued by his carer (1)e. Because of multiple daily visits from carers John similarly agreed that there were no benefits of telemonitoring for him, however he suggested 99% of people were likely to benefit: (1)f.

[Table 2 about here]

Reducing the Unnecessary

Given the accessibility of timely intervention via telemonitoring, participants were keen to utilise the device to reduce the current hospital appointments unless it was necessary. Five patients suggested Careportal® follow-ups to be more cost and time effective, besides reducing the hassles of attending the clinic – often described as brief with little benefit. In particular, James and Charlie, did not perceive any need for them to attend the clinics when little change in their physical well-being over time (2)a,b. Similarly, potential positive impact on health economics was noted by two participants. (2)c.

For those more physically impaired by the condition, reduced hospital appointments were perceived to be beneficial as these were stressful experiences. Similar to time/cost effectiveness, these views were in the context of perceiving little benefit of face-to-face consultations. (2)d. Face-to-face consultation, however, was not completely dismissed. Mary and Logan were keen to regularly attend hospital because of the perceived advantage of direct contact with a consultant. (2)e. Nevertheless, the benefits may depend on the usefulness of hospital clinics considering Mary also suggested reducing appointments elsewhere in her interview.

[Table 3 about here]

Increased self-awareness

Participants were supportive of the regular monitoring of symptoms required by telemonitoring. Patients reported that this was a feature of having MND – appreciating that there are symptoms to experience and cope with. Their acceptance of telemonitoring then appeared to be a consequence of patients' general understanding of fluctuations in their physical well-being (3)a.

Telemonitoring further enabled symptom awareness and interpretation for four patients. For one patient, this was through voluntarily taking extra readings of pulse oximetry: (3)b. For Logan, it was through answering symptom related questions (3)c. Yet for another patient, having a follow-up conversation with the physiotherapist increased the symptom awareness. Furthermore, it was pointed out by two relatives that regularly monitoring symptoms led patients to be less biased and to become objective about their condition: (3)d

As illustrated above, being accurate about their symptoms and coming to understand more about their physical well-being as a part of the process of reporting through the Careportal® was perceived to be beneficial, and this was also voiced by Mary herself and Charlie. These accounts and the following quote suggest that telemonitoring can promote an active engagement with self-care (3)e.

[Table 4 about here]

Taking Initiative

Five of the seven patients were positive about their experiences from using the message system on the Careportal® because it allowed them to get in touch with the healthcare providers when they needed support (4)a. As illustrated, patients were able to register their concerns as the need arose, with the assurance of future contact from the healthcare providers. And, even though the answers may not be instant, this gave the patient 'peace of mind'. The concerns that led to engaging with the

professionals were in relation to symptoms, general information, and equipment needs. This service format based around their needs was a sharp contrast to current communication with hospitals which was described as a one-way communication or communications with a long wait at best: (4)b.

Although the benefit of having the contact point was observed, this was not shared by William or John. Whilst William perceived the potential benefits, he had problems with sending messages (as shown below) therefore he never used the Careportal® to initiate contact. John did not perceive the benefits of having Careportal® as a contact point because he already had a good contact system set up as suggested by his carer: (4)c.

[Table 5 about here]

Technical challenges

Although participants expressed general ease of using the telehealth device, a few technical issues were reported. Whilst most challenges were perceived as minor, or they were quickly solved by the Careportal® provider, four participants expressed frustration with the messaging system – in particular on-screen keyboard. This was used to draft messages to their health providers. Participants said the keys were too small, as well as the touch panel having insufficient sensitivity and the time allowance for formulating a message was also insufficient. In particular, the on-screen keyboard issues discouraged William from using the message system: (5)a.

Other challenges were oximetry transmission (n=4), device fault (n=2), and mobile signal loss (n=1).

[Table 6 about here]

Discussion

Interviews with seven MND patients who had used a weekly telemonitoring service via Careportal® strongly suggest that they accepted telehealth as a suitable support for them, and that it may provide new benefits to MND healthcare provision. Regular assessments of physical well-being via Careportal® were considered not only beneficial to symptom management, but also to psychological well-being because of the perceived continuity of support by medical staff. Using telehealth to potentially reduce unnecessary cost, time and hassles of attending hospital appointments were identified as an important benefit. The telehealth approach led participants to better understand their condition and enabled them to actively seek support for their needs.

An investigation into the usefulness of telehealth in the care of people with MND has been slow compared to its development in other illnesses. To our knowledge, this was the first qualitative study to explore the experience of using remote monitoring for ventilated people with MND. The current study explored 'why' participants in the nurse-led telephone follow-up may have been satisfied with telehealth [7]. The findings of our study reflect the positive evaluations of people with other chronic conditions such as diabetes and chronic heart failure reported in previous qualitative works examining the subjective experience of telemonitoring. In line with these studies [12,13,18,19,24], patients suggested that monitoring their symptoms increased their knowledge of their symptoms – and this helped some individuals to establish their 'norm' [12,19]. In addition, as increased knowledge of one's own health status has been found to encourage some patients to engage with behavioural health management including treatment adjustment [12,15,24], we similarly found proactive symptom management. In our study, the utilisation of the Careportal® device increased self-awareness of well-being amongst participants which promoted an active engagement with self-care. The positive impact of telemonitoring on psychological well-being observed in the present study has been also supported by previous studies, reporting telemonitoring gives patients a sense of reassurance [12,13,16,18,25]. Due to positive experiences and also benefits

of regular monitoring leading to timely interventions, our participants preferred telemonitoring to hospital appointments if they perceived hospital visits to create more burdens than benefits. This favourable response to a possible reduction of clinic appointment echoes the patients' acceptability of telehealth in the care of MND by a recent study, which reported on the development of a telehealth system [11]. Similar to previous report [12,14,16,17,19], technical challenges of telemonitoring were generally perceived as a hassle, but did not detract from the benefits.

The current study also provides a novel account of an individual for whom there was no perceived benefit of telemonitoring. Previously, an ambivalent attitude towards the benefits of telemonitoring was expressed mainly by professional carers [26], and therefore this study adds to the current understanding on potential patients to whom telehealth may provide little benefit. One study reports professionals' view that telemonitoring is more beneficial for those with advanced condition with chronic heart failure than for those with stable symptoms [13]. In MND, initiation of NIV seems to be a good indication for introducing telemonitoring given the main cause of death is pulmonary complications [27,28]. Interestingly the current work notes that telemonitoring may not be perceived necessary for those with frequent daily visits from professional caregivers; though it should be noted the patient may not grasp the value of the technical data monitored and how that might be used to optimise care delivery such as ventilator settings.

Limitations of the current study

No interview was conducted with patients who had declined to be part of the Telehealth trial that preceded this research [5]. Given that one of the factors affecting the uptake of telehealth is people's motivation [29], perspectives of such individuals would be useful to scope if telehealth was to be translated into a formal service approach. Also, we have not sought interviews with the healthcare providers to explore their experiences of utilising the telemonitoring to provide care for ventilated patients with MND. Previous studies on telehealth in MND reported time and cost effectiveness of telehealth [7-10], yet studies in other chronic conditions found difficulties of

information sharing amongst professionals [13,14,23,26,30] which was potentially time consuming. Given the importance of multidisciplinary teams in delivering care in MND, the aspect of communication difficulties, and other potential problems, of using telehealth amongst the professionals needs to be defined. Although we attempted to capture the experience of lay carers in our study, only joint interviews were conducted at the request of both patients and caregivers. Nevertheless, it is reflected that carers of people with MND experience great burdens due to the illness [31], and thus a separate interview may have provided an environment for open discussion. Further examinations of carers' experience of using telehealth in the care of MND NIV patients would be crucial especially in situations where patients become completely dependent on carers.

Conclusions

The findings of this study suggest that telemonitoring via Careportal® may facilitate delivery of NICE recommendations relating to the care for people with MND. It is anticipated that this would extend to similar systems with appropriate procedures. In particular, amongst the non-invasively ventilated patients we studied here, telemonitoring did not compromise patients' experience of healthcare services, and for six of our seven participants it was enhanced. Telehealth enables clinicians to remotely monitor patients, thus no travel is required, thereby providing an alternative to support those who cannot attend the clinic. It may be argued, therefore, that regular assessment through telemonitoring may reduce clinical appointments while promoting tailored care fitting the individual's needs. The increased awareness of symptoms through telemonitoring is further aided by the messaging system, which functions as a contact point to appropriately notify clinicians of the issues and the need for further assessment. This is particularly beneficial when a patient experiences a significant change in symptoms. It was only here where full appreciation of motor challenges for this patient population is needed to ensure there are no barriers with being able to send a message. Effective interactions made through telehealth are anticipated to also contribute to efficiency savings if, this became a general approach. This fits well with the NHS Quality, Innovation,

Productivity and Prevention (QIPP) approach which is designed to improve the quality of care, while making efficiency savings to reinvest into the system [32].

The finding of positive experiences of using telemonitoring reported here is encouraging as it suggests that telemonitoring, using a device such as Careportal®, would allow an effective tailored approach while enhancing the experience of patients and their caregivers. The results advocate further investigation, ideally with a controlled trial, and separate caregiver interviews, to assess the overall impact of telehealth on care in MND patients, particularly those in the advanced stage on NIV. We are now designing a study to test the feasibility of telemonitoring for the routine care of NIV MND patients in collaboration with a local Clinical Commissioning Group (CCG) which provides telehealth to other illness groups [5].

References

1. National Institute for Health and care Excellence. Motor neurone disease: assessment and management, https://www.nice.org.uk/guidance/ng42 (2016, accessed 6 June 2019)

2. Chio A, Bottacchim E, Buffa C, et al. Positive effects of tertiary centres for amyotrophic lateral sclerosis on outcome and use of hospital facilities. *J Neurol Neurosurg Psychiatry* 2006; 77: 948-950.

3. Talbot K. Motor neurone disease. *Medicine* 2004; 32: 105-107.

4. Neudert C, Oliver D, Wasner M, et al. The course of the terminal phase in patients with amyotrophic lateral sclerosis. *J Neurol* 2001; 248: 612-616.

5. Anonymous, 2019.

6. Selkirk SM, Washington MO, Mcclellan F, et al. Delivering tertiary centre specialty care to ALS patients via telemedicine: a retrospective cohort analysis. *Amyotroph Lateral Scler Frontotemporal Degener* 2017; 18: 324-332.

7. Vitacca M, Comini L, Tentorio M, et al. A pilot trial of telemedicine-assisted, integrated care for patients with advanced amyotrophic lateral sclerosis and their caregivers. *J Telemed Telecare* 2010; 16: 83-88.

8. Vitacca M, Comini L, Assoni G, et al. Tele-assistance in patients with amyotrophic lateral sclerosis: long term activity and costs. *Disabil Rehabil Assist Technol* 2012; 7: 494-500.

9. Pinto A, Almeida JP, Pinto S, et al. Home telemonitoring of non-invasive ventilation decreases healthcare utilisation in a prospective controlled trial of patients with amyotrophic lateral sclerosis. *J Neurol Neurosurg Psychiatry* 2010; 81: 1238-1242.

10. De Almeida JPL, Pinto A, Pinto S, et al. Economic cost of home-telemonitoring care for BiPAP-assisted ALS individuals. *Amyotroph Lateral Scler* 2012; 13: 533-537.

11. Hobson EV, Baird WO, Partridge R, et al. The TiM system: developing a novel telehealth service to improve access to specialist care in motor neurone disease using user-centered design. *Amyotroph Lateral Scler Frontotemporal Degener* 2018; 19: 351-361.

12. Fairbrother P, Pinnock H, Hanley J, et al. Exploring telemonitoring and self-management by patients with chronic obstructive pulmonary disease: a qualitative study embedded in a randomized controlled trial. *Patient Educ Couns* 2013; 93: 403-410.

13. Fairbrother P, Ure J, Hanley J, et al. Telemonitoring for chronic heart failure: the views of patients and healthcare professionals – qualitative study. *J Clin Nurs* 2014; 23: 132-144.

14. Hanley J, Ure J, Pagliari C, et al. Experiences of patients and professionals participating in the HITS home blood pressure telemonitoring trial: a qualitative study. *BMJ Open.* [Internet].

2013 May 23 [cited 2019 Jun 6]; [10 p.]. Available from https://bmjopen.bmj.com/content/3/5/e002671

15. Hanley J, Fairbrother P, McCloughan L, et al. Qualitative study of telemonitoring of blood glucose and blood pressure in type 2 diabetes. *BMJ Open.* [Internet]. 2015 Dec 23 [cited 2019 Jun 6]; [8 p.]. Available from https://bmjopen.bmj.com/content/5/12/e008896?utm_source=trendmd&utm_medium=cpc&utm_campaign=alljjs&utm_content=americas&utm_term=1-B

16. Hanley J, Fairbrother P, Krishan A, et al. Mixed methods feasibility study for a trial of blood pressure telemonitoring for people who have had stroke-transient ischaemic attack (TIA). *Trials* 2015; 16: 117. doi: 10.1186/s13063-015-0628-y

17. Pekmezaris R, Schwartz RM, Taylor TN, et al. A qualitative analysis to optimize a telemonitoring intervention for heart failure patients from disparity communities. *BMC Med Inform Decis Mak* 2016; 16: 75. doi: 10.1186/s12911-016-0300-9

18. Penica JL, Vickers KS, Finnie DM, et al. Telemonitoring increases patient awareness of health and prompts health-related action: initial evaluation of the TELE-ERA study. *Telemed J E Health* 2011; 17: 461-466.

19. Riley JP, Gabe JPN and Cowie MR. Does telemonitoring in heart failure empower patients for self-care? A qualitative study. *J Clin Nurs* 2012; 22: 2444-2455.

20. Cedarbaum JM, Stambler N, Malta E, et al. The ALSFRS-R: a revised ALS functional rating scale that incorporates assessments of respiratory function. *J Neurol Sci*. 1999; 169: 13-21.

21. Montes J, Levy G, Albert S, et al. Development and evaluation of a self-administered version of the ALSFRS-R. *Neurology*. 2006; 67: 1294-1296.

22. Chamberlain K. Methodolatry and qualitative health research. *J Health Psychol* 2000; 5: 285-296.

23. Braun V and Clarke V. Using thematic analysis in psychology. *Qual Res Psychol* 2006; 3: 77-101.

24. Inglis SC, Clark RA, Dierckx R, et al. Structured telephone support or non-invasive telemonitoring for patients with heart failure. *Cochrane Database Syst Rev.* [Internet]. 2015 Oct 31 [cited 2019 Jun 6]; [256 p.]. Available from https://www.cochranelibrary.com/cdsr/doi/10.1002/14651858.CD007228.pub3/abstract

25. Fairbrother P, Pinnock H, Hanley J, et al. Continuity, but at what cost? The impact of telemonitoring COPD on continuities of care: a qualitative study. *Prim Care Respir J* 2012; 21: 322-328.

26. Elwyn G, Hardisty AR, Pierce SC, et al. Detecting deterioration in patients with chronic disease using telemonitoing: navigating the 'trough of disillusionment'. *J Eval Clin Pract* 2012; 18: 896-903.

27. Andrew J. Amyotrophic lateral sclerosis: clinical management and research update. *Curr Neurol Neurosci Rep* 2009; 9: 59-68.

28. Winhammer JMC, Rowe DB, Henderson RD et al. Assessment of disease progression in motor neuron disease. *Lancet Neurol* 2005; 4: 229-238.

29. Greenhalgh T, A'Court C and Shaw S. Understanding heart failure; explaining telehealth – a hermeneutic systematic review. *BMC Cardiovasc Disor.* [Internet]. 2017 Jun 14 [cited 2019 Jun 6]; [16 p.]. Available from https://bmccardiovascdisord.biomedcentral.com/articles/10.1186/s12872-017-0594-2

30. Peirce SC, Haristy AR, Preece AD, et al. Designing and implementing telemonitoring for early detection of deterioration in chronic disease: defining the requirements. *Health Inform J* 2011; 17: 173-190.

31. Aoun SM, Bentley B, Funk L, et al. A 10-year literature review of family caregiving for motor neurone disease: moving from caregiver burden studies to palliative care interventions. *Palliat Med* 2013; 27: 437-446.

32. Department of Health. *Equality and Excellence: Liberating the NHS*. London: Stationery Office, 2010.

Table 1: Demographics and illness characteristics of patients at the time of trial

Patient (pesudonym)	age	Illness duration	NIV duration (month)	Onset type	ALSFRS-R Baseline	ALSFRS-R Post-trial	Carer	Living arrangement
Charlie	62	11y 10m	18	Limb	21	21	Spouse	with a spouse and child(ren)
James	58	5y 7m	34	Limb	37	31	-	with a partner
John	49	13y 7m	36	Limb	6	6	Professional home carer	alone
Logan	66	14m	12	Respiratory	37	28	Spouse	with a spouse and child(ren)
Margaret	68	8m	0	Limb	25	15	Daughter	alone
Mary	68	14m	10	Limb	22	21	Spouse	with a spouse
William	71	7m	4	Limb	34	33	-	with a child(ren)

NIV: non-invasive ventilation, ALSFRS-R: revised amyotrophic lateral sclerosis functional rating scale [20,21]

Table 2: Quotes for 'Benefits of timely intervention'

Quote	
(1) a	You know that was very good. Because the oximetry, when it worked, it was very good because [physio] could be on the work: "Well, we need to alter these settings [on NIV]". You had that once, didn't you? [Mary's husband]
(1) b	I don't see myself as being that ill. But I can really understand needing it or using it if you are in the worse situation sort of thing. You could have poor breathing because of a disease – MND – or whatever. I could see it being really useful then. [James]
(1) c	It was probably just making me feel a little bit safer because I knew when you… get all the answers, as soon as you've seen something not regular, you'd ring me up. [Logan]
(1) d	Once it starts to deteriorate more it would be even more… you know, more vital to have it [Careportal®] then. Because if you are okay up here, your well-being is going to be better. You know, [if] you are thinking more; if you are worrying about everything then it's going to be worse for you. [Charlie]
(1) e	Because he [John] has those carers coming in like five times a day, six times a day – I don't know. Six times a day? [John indicates six] It is [six]. You know morning and then the mid-morning like now and there's lunch call and then it's mid-afternoon, then it's tea call, then it's mid-evening call, and then put to bed call. So, he's always got somebody in here anyway and we should all notice if anything happens to him. [John's home carer]
(1) f	It [Careportal®] would be handy for 99% of people. [John]

Table 3: Quotes for 'Reducing the unnecessary'

Quote	
(2) a	I go to [hospital 1] every six months with [neurologist] and that [it] lasts for between 30 seconds and 2 minutes. He goes, 'How are you? Yeah, yeah. Okay. See you then.' And the same [physician] I've probably seen – is it every 6 months? Or 12 months? I think it's 6 months I see [physician] or one of his people. Same sort of thing – I walk in and 'How are you?' 'Yeah, fine off you go.' If it stops you doing that it would be brilliant. There is no need. [James]
(2) b	It'll save a lot of money as well as you know because I won't need to keep going to hospital when I don't need to go. [Charlie]
(2) c	It's – I'm taking their [clinicians] time up, taking car parking space up – it's just… Careportal® is the way to go for me [James].
(2) d	**Margaret:** [on iPad]: Doctor doesn't say much. **Daughter:** [Laughs] I know I know what she is referring to. I know. You see, when we go to the neurology clinic … Like I say, it's a big thing to get my mum washed, dressed, in a wheelchair to the hospital and you sort of go in and 'How are you?' 'Alright' 'Okay then I will see you.' It's literally that short that appointment. There's no - there's no benefit to her do you know? It's a big thing for my mum to go there do you know what I mean? It's like, 'Why?' [Laughs] You know so in that respect yeah because it… there is no point in going to that appointment do you know what I mean because you get absolutely nothing – not a thing.
(2) e	I would like to see the doctors now and again just for peace of mind. […] They tell you exactly what you should be doing or what you are doing wrong and even if there's a slight deterioration you know they tell you. [Logan]

Table 4: Quotes for 'Increased self-awareness'

Quote	
(3) a	Basically, I'm monitoring every day in my head anyway. I know when I'm having a good day and when I'm having a bad day. [Logan]
(3) b	But sometimes you can feel … and you think, 'I'll check that'. Then you know yourself when you are not quite up to scratch then it [oximetry reading] will just confirm it – that's about it. [Charlie]
(3) c	When you get questions on about 'Have you had phlegm?' and 'What colour the phlegm is?' and all that - it probably makes you think about that side of it because without pressing them buttons, I wouldn't even think about that. [Logan]
(3) d	I think it is very easy for people to just get locked into what they are experiencing, and you can look at it in a more negative ways, whereas the Careportal® was asking questions and you think, 'Oh… I answered similar earlier in the week', you know, 'Things are ok.' It's not, 'Oh no, I feel a lot worse', or whatever. Or, 'I'm having more and more phlegm' – I think it was just making us both aware of what was happening. And it was fairly consistent. [Mary's husband]
(3) e	**Patient:** I think you've got to be honest with your answers. **Interviewer**: So, if you were not completing the Careportal®, you might just ignore symptoms? **Patient:** There was – 'Have you had phlegm?' Maybe you had a little bit [but] you put 'No'. So, not helping yourself.

Table 5: Quotes for 'Taking initiative'

Quote	
(4) a	If you've got a problem, no matter what time it is, you can type it in. Okay, there is no-one to be there until the next day, but you type in what your problem is at that time and you know at some point someone will get back to you. [James]
(4) b	We don't really have a great deal of communication. The only thing you have is they send you out an appointment and you go to your appointment and you come away until next one and that's pretty much all the communication that you have. [Margaret's daughter]
(4) c	Very rare, isn't it that you [John] need to speak to anybody, is there really? And if you do, you'll send an email to somebody, won't you? You know, he'll… that's how he'll contact people - by an email. [John's home carer]

Table 6: Quotes for 'Technical challenges'

Quote	
(5) a	The keyboard is that frustrating I just couldn't be bothered trying to get it to work because it wouldn't. [William]

Interview schedule for semi-structured interviews

Both patient and caregiver:

1. What was it like to use the Careportal?
- Positive
- Negative

2. Was the Careportal useful at that time it was offered to you/patient? (Ask participants to expand their response.)

3. Were there any problems resulted from using the Careportal? If so what were they?

4. Please tell us any bad points about using this device. (if not already covered in Q1)

5. Did you have any contacts from clinicians due to the answers you provided on the device? How useful was the follow up action?

6. How did you find this way of working with you in comparison to the normal way of treatment you receive?
-before Careportal
-after Careportal

7. Was there any time you wanted to get in touch with us using Careportal, but you didn't? If so, what stopped you?

8. Do you think using this device in MND service routinely would be useful and if so why/how?

9. How did you feel about monitoring your own/patient's symptoms of MND? (positive/negative)

10. How did you find the handling of the device?

11. How can we improve technical aspects of Careportal?

Additional question for caregiver (after the first question):

12. What impact did the Careportal have on you as opposed to its impact on [Patient]?

Appendix 2: Qualitative codes

Theme	Supreme code	Code	Source (no. of participants): name
Benefits of timely Intervention	Effectiveness of telemonitoring	Real time information	6: James, Charlie, Mary, Logan, William, Margaret
	Physiological well-being	Effective interactions	5: James, Charlie, Mary, William, Margaret
	Psychological well-being	Enjoy using Careportal®	2: Charlie, Mary
		Feeling Valued	1: Mary
		Good feeling	1: William
		Peace of mind/sense of security	5: Charlie, James, Mary, Logan, Margaret
		Significance of psychological well-being	2: Charlie, Mary
	No perceived benefits	No perceived medical benefits	3: James, Charlie, John
		Already having a good support system	1: John
Reducing the Unnecessary	No perceived benefits of outpatient appointment		5: James, Charlie, John, William, Margaret
	Savings	Health economics	2: James, William
		Hassles of hospital visits	3: Mary, William, Margaret
		Less financial burdens	1: Charlie
	Value of face-to-face consultation		4: James, Charlie, Mary, Logan
Increased Self-awareness	Increased Self-awareness	Self-monitoring	4: Charlie, James, Mary, Logan, Margaret
		Careportal® as educational	2: Charlie, Logan
	Further promoting self-care		2: Charlie, Mary
Taking initiative	Active role in the care	Message to get in touch	5: James, Charlie, Mary, Logan, Margaret
		Careportal® as a backup	5: James, Charlie, Mary, Logan, Margaret
	Contact with hospital	Asking questions via messaging system	3: Charlie, Mary, Margaret
		Contact to get accessories	2: Charlie, Logan
		Personal contact through Careportal®	2: Mary, Logan

		Traditional one-way communication with hospital	4: Charlie, Mary, John, Margaret
Technical Challenges	Easiness	Easiness of Careportal®	5: James, Charlie, John, Logan, Margaret
	Technical issues	Messaging system	4: James, Mary, Logan, William
		Entry time	3: Charlie, Mary, William
		Faulty machine	2: Logan, Margaret
		Oximetry related	5: James, Mary, John, William, Margaret
		Signals	1: Charlie